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Research Letter

Benign metastasizing leiomyomas thought to be nodal metastases in a case of ovarian cancer

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Dear Editor,

Uterine leiomyomas are the most common gynecologic tumors in women of reproductive age and represent the most common indication for hysterectomy in premenopausal women. In rare instances, these tumors occur outside of the uterine corpus and are referred to as 'benign metastasizing leiomyomas' (BMLs) [1].

A 51-year-old nulligravid postmenopausal female without a history of gynecologic problems or prior surgeries presented with dyspnea. Computed tomography (CT) showed a 13.3-cm pelvic mass, peritoneal carcinomatosis, retroperitoneal lymphadenopathy, and a large pleural effusion. Her CA-125 was 8764 μ mL. An excisional biopsy of a palpable left supraclavicular lymph node was consistent with adenocarcinoma favoring Mullerian origin. Given the extent of disease seen on imaging, it was felt optimal cytoreduction would not be possible. She therefore underwent four cycles of neoadjuvant paclitaxel and carboplatin, and her CA-125 decreased to 26 μ mL. A post-treatment CT scan showed a cystic left ovary, myomatous uterus, and a small pleural effusion, but no lymphadenopathy or ascites.

The patient therefore underwent an interval debulking surgery. On vaginal exam, she had a uterus consistent in size with a 14-week gestation, irregular and firm palpable masses bilaterally, and palpable nodularity on rectovaginal exam. Intraoperatively, she had bilateral 4-cm ovarian masses densely adherent to the rectum and bilateral pelvic sidewalls, two 1-cm omental implants, and enlarged right paraaortic lymph nodes. In order to achieve no gross residual disease, it was necessary to perform an extended total

abdominal hysterectomy with lysis of adhesions, bilateral salpingo-oophorectomy, omentectomy, and right paraaortic lymph node dissection. Pathology was consistent with high-grade serous carcinoma in the left ovary, bilateral fallopian tubes, and omentum (Fig. 1). BMLs were noted in the right parametria and in three of the four paraaortic lymph nodes. There was no cytologic atypia, necrosis, or increased mitotic activity (Fig. 1). Immunohistochemical stains were notable for 100% nuclear staining for estrogen and progesterone receptors.

Uterine smooth muscle tumors with recurrent or metastatic potential include BML, intravenous leiomyomatosis, and uterine smooth muscle tumor of uncertain malignant potential and must be distinguished from leiomyosarcoma [2]. While the majority of BMLs occur in the lungs, they have also been found in the abdomen, lymph nodes, deep soft tissue, heart, bone, and central nervous system [1,3,4]. Investigators have postulated that BMLs represent multifocal smooth muscle proliferation attributable to coelomic metaplasia or that they originate from microscopic vascular invasion of uterine leiomyomas [1]. This latter theory is supported by the observation that BMLs are predominantly seen in women with antecedent myomectomies or hysterectomies for uterine leiomyoma, often in the remote past [4].

BMLs generally have an indolent course, but occasionally may become extensive or metastasize to key structures, causing significant morbidity or mortality [5]. Reported treatment modalities include careful observation, surgical resection, progestins, aromatase inhibitors, GnRH agonists, and bilateral salpingo-oophorectomy. Particularly in premenopausal women, surgical resection of metastatic lesions does not guarantee remission. Surgical or medically induced menopause has successfully stabilized the disease or induced remission [3]. Our current knowledge of BMLs is derived from single-institution case series. Therefore, further research should seek to better characterize the etiology of BML, distinguish BML from other smooth muscle neoplasms, predict those at risk for further disease progression, and tailor treatment to minimize further morbidity.

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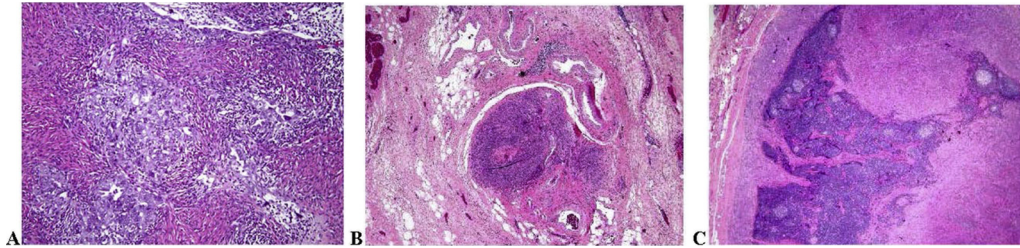


Fig. 1. Morphological features of the ovarian tumor and leiomyoma (hematoxylin and eosin stain). A: Ovarian high-grade serous carcinoma with marked nuclear atypia and pleomorphism (200 \times); B: Benign metastasizing leiomyoma involving parametrial soft tissue and paraaortic lymph node (C) (20 \times).

Our patient presented with widely metastatic ovarian cancer thought to involve her paraaortic lymph nodes. Although this did not change her prognosis or treatment, it is interesting to note that the bulky lymph nodes were a benign neoplasm. There may be other instances in which physical exam, imaging, or operative findings concerning for malignancy but found to be BML may change a patient's prognosis and potentially treatment. Due to their recurrent and metastatic behavior, imaging and intraoperative assessment of BMLs must be confirmed with pathologic diagnosis.

Authorship statement

All authors contributed equally to the drafting and editing of the manuscript. Dr. Hussein generated the images that demonstrate the tumor histology.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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